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Case series of Fetus in fetu- Variable presentation

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ABSTRACT

Background: Fetus in fetu is a rare congenital anomaly. It comprises of a malformed twin incorporated into the surviving twin.

Materials and method: Diagnosis is made preoperatively through ultrasonography, plain radiography, computed tomography or MRI. Histopathology confirms the diagnosis; here we are reporting a series of four cases of fetus in fetu.

Conclusion: Treatment is en-bloc excision. Histopathology will lead to further line of treatment.

Keywords: *Fetus in fetu, Teratoma, abdominal mass, congenital anomaly, infants.*

1. INTRODUCTION

Fetus in fetu (FIF) is a rare congenital anomaly. Historically it can be traced back to the German Anatomist-Johann Meckel for describing as early as 18th century. It is comprised of a malformed twin incorporated into the surviving twin usually presenting as an abdominal mass. The pathophysiology and differentiation of FIF from teratoma is well explained; the primary distinction between mature teratoma and FIF is that the latter has the capacity for autonomous growth and malignant potential.[1] Its incidence is 1 in 500,000 births, and the majority of cases occur in infancy.[2] Diagnosis is made preoperatively through various imagining techniques. Histopathology confirms the diagnosis and treatment is excision. The majority of cases have been reported in retroperitoneum (80) %.

2. CASE REPORTS-

Case Report- 1

A 5-month-old female child presented with gross abdominal distension with respiratory distress. It had a history of abdominal mass since birth which was gradually progressive up to presentation. The child was born through caesarean section at full term without any complications. None of the family members had a history of twin births. On inspection gross abdominal distension was noted. By doing per abdominal examination, it was noted that a firm well well-defined spherical mass could be palpated occupying whole of the upper and lower quadrant of right side of the abdomen crossing the midline.

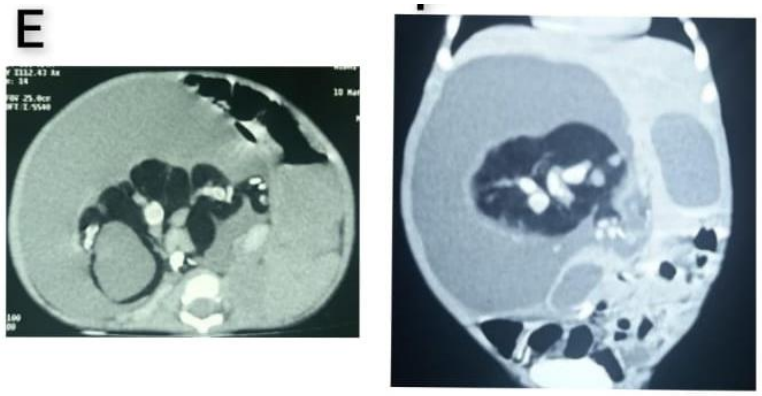
All the routine blood investigations were under normal limits. Serum β-HCG, CEA, and LDH were under normal limits. However, Serum AFP levels were found to be elevated. On Ultra Sonography of abdomen, a large cystic mass with internal solid components shows an orbit-like structure, limb buds in the right side of the abdomen in the

retroperitoneum pushing the bowel loops anteriorly. Contrast CT of Abdomen revealed a well-encapsulated multiloculated solid cystic lesion measuring ~112 x 136 x 141 mm (AP x TR x CC) with intralesional fatty component and calcification with formation of appendiceal bones in right hemi abdomen crossing midline and arising from the retroperitoneum. [Image-1]

Thus, preoperatively fetus in fetuses and highly organized teratoma were kept as differential diagnoses. But formation of appendiceal bones with limb buds and orbit-like structures goes more in favor of fetus in fetu.

Exploration by laparotomy was done which revealed a large cystic mass in the retroperitoneum. The cystic encapsulated mass was excised in toto after careful dissection and ligation of feeding vessels. The baby did well with the treatment and had been discharged on the 10thpostoperative day.

On gross examination, a well-encapsulated mass with solid and cystic components measuring around 16 x 18 x 14 cm was found. The solid component consisted of malformed limbs with vestigial fingers, hair, skin, and orbit-like projections. On cutting transversely through the glistening surface, well-developed vertebral columns were also identified. [Image- 2]



CECT Abdomen- well-encapsulated multiloculated solid cystic lesion.

[image- 2]

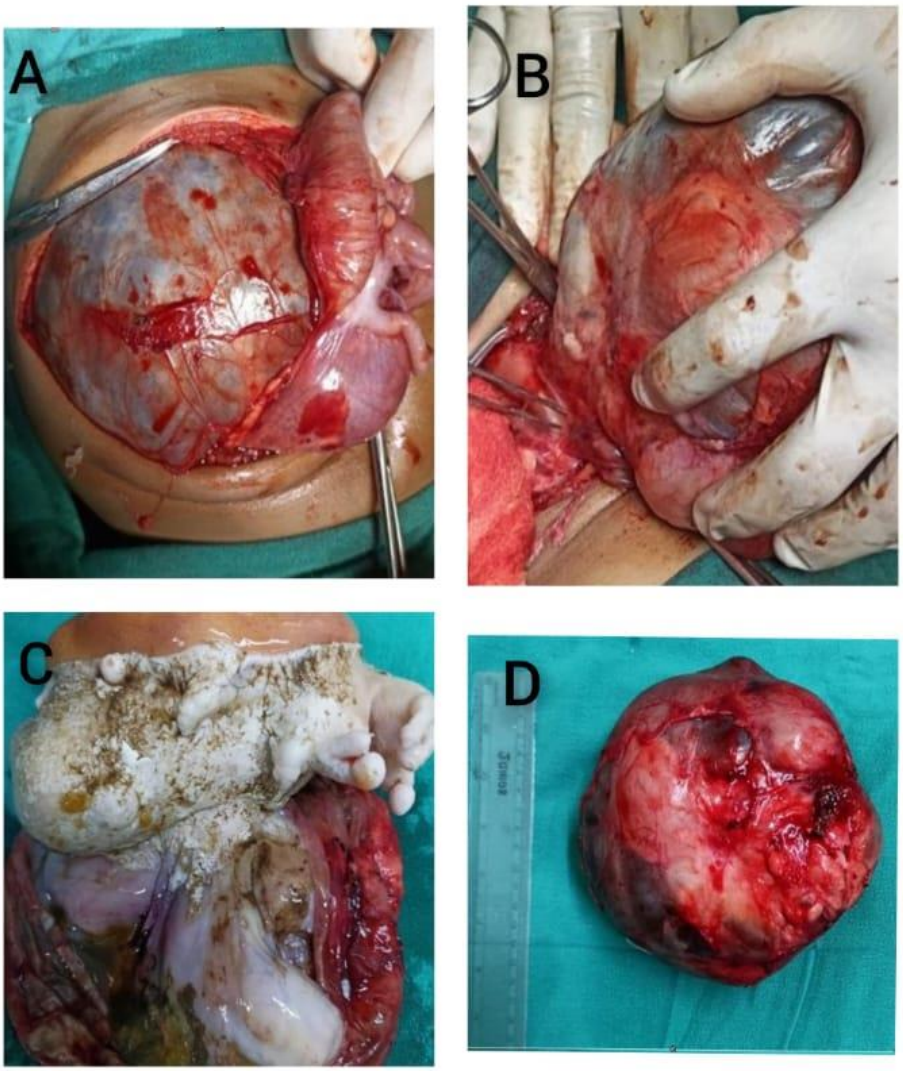


Image- 2 intraoperative findings

Sections of the cyst wall seen under a microscope revealed fibro collagenous tissue lined by the squamous epithelium. Sections from the fetus showed the presence of intestinal, cartilaginous, and glial tissue. Sections from the vertebral column show the presence of marrow elements, bone, and fibrocartilaginous tissue.

Case Report-2

We here present diagnosis and management of a case of Fetus In fetu in a three years, female child weighing 10 kgs. There was no history of maternal sickness, radiation exposure, or drug use during pregnancy, and neither parent reported twinning or congenital abnormalities in previous deliveries. It was a fixed firm nontender mass in the right gluteal region. Blood investigations were within normal limits.

Initial imaging with plain abdominal film revealed calcified densities and ultrasonography and abdominal computed tomography revealed a large mass extending from lower abdomen to the pelvic region. It had cystic and solid calcified components with a pressure effect on bladder giving a differential diagnosis of sacrococcygeal teratoma.

Mass was approached through perineal incision; abdominal part was mobilized from there only. On table, the mass was found to be cystic and solid. The mass was completely excised. On cut section the capsule was a feti form structure, one upper limb was well developed, and structures resembling fingers were rudimentary leading to the diagnosis of FIF.

Case report-3

LSCS delivered a full-term boy weighing 3.5 kg. No relevant history. A complex cystic and solid abdominal mass was

discovered during an antenatal ultrasound conducted at 30 weeks of gestation. Distention was discovered during a postnatal abdominal check. Upon examination, a 7.5 × 5.5 cm tumor was felt in the upper left abdomen. Every tumor marker was within normal limits. A postnatal abdominal radiograph confirmed the mass. It was identified as a well-encapsulated mass retroperitoneal, anterior to the left kidney, by abdominal ultrasonography and CT scan. Since all of the imaging modalities displayed bony prominence, they were all displaying characteristics that were favorable to the fetus. This served as a FIF diagnostic. After an elective laparotomy, a 7.5 × 6.0 x 6.0 cm mass was effectively removed. The weight of the capsule was 120g. The spinal column was perceptible after the capsule was cut. The FIF possessed two upper extremities, with four fingers on one hand and five on the other. In terms of histopathology, the FIF revealed skin, intestines, and skeletal muscles. The time following surgery was uneventful.

Case report- 4

A one-month-old male child presented with a normal antenatal scan with adequate liquor with postnatal ultrasonography finding of a heterogenous cystic lesion noted in left retroperitoneum extending from spleen to pelvis, posterior and medial to left kidney of size 62*21 mm which also contain an echogenic lesion having size 22*22 with internal vascularity and calcification impression suggestive of retroperitoneal teratoma, Patient was advised CECT.

CECT abdomen which was suggestive of a well encapsulated multiloculated solid cystic enhancing lesion measuring-45*68*93mm (AP*TR*CC) with intralesional fatty component calcification appearing as if appendiceal bones, differential diagnosis included fetus in fetu and teratoma. All the blood investigations were with in normal limits. A retroperitoneal mass measuring approximately 5*7*6 cm was removed during surgery, and it was sent for histopathological analysis. The specimen was grey-white with fine hair when it was cut, and the cystic surface showed pearly white cartilaginous areas, firm white vague nodular appearances, The specimen's microscopic description showed that it contained haphazardly distributed mature tissue derived from ectoderm, mesoderm, and endoderm. The vertebral column and limbs were identified, and the specimen's morphology was favourable to the fetus.

3. DISCUSSION

Fetus in fetu and teratoma is a diagnostic dilemma. In our cases, the site of the mass is retroperitoneum and sacro-coccygeal which is one of the most common sites of FIF, as well as presence of limb buds, vestigial fingers, orbit-like structures, intestinal components, glial cells, and vertebral elements help in distinguishing fetus in fetu from teratomas. Whereas teratomas in terms of site, are mostly present in the sacrococcygeal, ovaries, and lower part of the abdomen

In 1953 Willis emphasized that FIF is a mass having a vertebral column along with different organs or limbs. Our cases meet this criterion very well. Among the differential diagnoses include teratoma, meconium pseudocyst, FIF, and neuroblastoma.[2]Prior to the advancement in imaging techniques preoperative diagnosis of FIF has been made in only 16.7% of cases. Sonography and a CT scan were useful in our situation for preoperative evaluation in order to suspect a fetus; as a result, an early diagnosis with better results is now possible[2]. As CT and MRI are used more frequently, the differential diagnosis window is now much smaller, resulting in a perfect diagnosis.[3] However, it was intraoperative findings and specimen examination that led to a confirmative definitive diagnosis. Therefore, surgical excision is the suggested course of treatment for FIF. Because this uncommon disease was only verified in our case following an exploratory laparotomy and examination of the excised specimen.

Due to a few instances of cancer after FIF resection, some surgeons have decided to do a more prompt complete resection and then follow up with a two-year postoperative evaluation of tumor markers[5]. AFP (Alpha-fetoprotein) and hormone HCG (human chorionic gonadotropin) are the two tumour markers which have to be kept in follow-up. There has been a lot of confusion in differentiation of FIF from teratomas[6].According to a case report by Hopkins et al., a 5-day-old boy with a retroperitoneal FIF in histology later developed a right abdominal tumor that was later identified as a teratoma with malignant components that needed chemotherapy[6]. Even though these cases are uncommon, they demonstrate the value of long-term monitoring for surveillance and to identify any tumors that would go undetected after surgical resection.

Conclusion- Complete excision (in-toto) is the treatment for fetuses. Histopathology can only result in additional therapy options. Reports of these uncommon instances, which describe the pathology, genetic findings, demography, and outcomes of this uncommon tumor, have increased in regularity.

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